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PRIAPISM FROM HYPERNEPHROMA METASTASES IN THE CAVERNOUS BODIES*

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IT is not my purpose to present a discussion of priapism. The paper is chiefly inspired by an unusual case of enlargement of the penis due to metastatic carcinoma of the erectile tissue, with priapism a secondary factor. Indeed, as in most similar cases, the priapism was incomplete; for the enlargement of the penis, and engorgement of the venous spaces, was not accompanied by muscular contraction nor the erect position of the organ. The term "false priapism," used by Gadrat, seems somewhat more appropriate.

LITERATURE

Scheuer,[1] in 1911, and Hinman,[2] in 1914, published excellent articles on priapism, and each in his classification included local malignant disease as one of the causes. In his tabulation of 133 cases from the literature, Scheuer mentions one of primary angiosarcoma, or endothelioma, reported by Maurer [3] in 1883, and one of metastatic carcinoma of the cavernous bodies from a primary carcinoma of the bladder published by Neumann [4] in 1882. He also mentions that Weber had a patient with metastases to the erectile tissue from a tumor of the testicle; but his catalogue of the cases and reference to Weber's [5] original article fail to reveal the case, and I have found no other reference to it.

In his review of 170 cases of priapism collected from the literature, Hinman found thrombosis of the veins of the penis in 125, and says that the venous thrombosis in two of these was due to local newgrowths. A study of his bibliography reveals that these were the same cases cited by Scheuer. It is obvious, therefore, that neoplasm, either primary or metastatic, is not a common cause of priapism.

COMMENT

It is true that primary tumors of the penis may sometimes invade the erectile tissue diffusely, causing enlargement of the entire organ and even priapism. Young [6] has pointed out that carcinoma of the glans, though it usually extends along the lymphatic channels to the regional lymph glands, may occasionally infiltrate the cavernous bodies by direct extension. Likewise, cancer of the urethra has been found to produce a similar result. Allenbach,[7] in 1916, described such an occurrence, and Culver and Forster [8] had a patient in whom the whole shaft of the penis became an indurated mass, through invasion, however, of the corpus spongiosum.

Malignant tumors primary in the cavernous bodies usually belong to the sarcoma group. Joelson,[9] in 1924, collected 34 such cases, and Cecil,[10] writing in Cabot's Urology in 1936, brought the number to 47. Of these, 13 were endotheliomata, of which, according to Joelson, the majority spread diffusely through the cavernous bodies and produce some degree of priapism. Of the other types of sarcoma, the melanoma practically never, and the others only rarely, infiltrate widely, but remain as a localized rounded nodule. However, the case presented by Frontz and Alyea,[11] in 1927, apparently was a primary sarcoma of the erectile tissue, with extensive metastases, and associated with a definite priapism. The account, as published, is not clear as to the origin of the tumor, and it has been referred to by other writers as a primary sarcoma of the prostate with metastases, but study of the evidence seems rather to favor a primary tumor.

The patient whose history I am presenting belongs to the smaller group in which the enlargement of the penis was due to invasion of the corpora cavernosa by malignant cells from a distant primary focus.

REPORT OF CASE

Mr. L. H., a man of 72, was admitted to the hospital on September 15, 1938, with a hematuria of three weeks' duration, much more severe in the last 24 hours. There had been fleeting pain in the region of the left kidney, slight frequency of urination, but no difficulty, though he had passed a few small clots.

His past medical history was quite extensive, and included several operations. He had been discharged from the hospital only a month before, after an illness of several weeks, characterized by high fever and rather severe right abdominal pain, for which no exact diagnosis had been made. While in the hospital numerous urinalyses had been made, all of which had been normal.

The physical examination revealed a tall, undernourished, elderly white male. Temperature, pulse, and respirations were normal. Blood pressure was 110/70. Loose-fitting, artificial teeth showed the marked loss of weight from his chronic illness. Heart and lungs indicated nothing of note. Abdomen revealed two operative scars, one with a large hernia. Both kidneys were palpable, the right easily so, as it was distinctly lower than normal. Neither kidney was tender nor nodular. Prostate was slightly enlarged and smooth. External genitalia at this time were entirely normal.

The blood count showed a moderate secondary anemia and a definite leukocytosis, with 24,000 white cells, of which 87 per cent were polymorphonuclears. Urine contained small clots, was distinctly red, and microscopically was loaded with erythrocytes and only a few leukocytes.

Cystoscopy was done the same day. The instrument passed easily and about one ounce of bloody urine was found in the bladder. A few small clots were aspirated. There was moderate prostatic obstruction, but no trabeculation of the bladder wall, and no tumors, stones, or foreign bodies. The urinary jets from the right ureteral orifice were clear, but those from the left side were distinctly bloody. The function of the right kidney was normal, while that of the left was reduced about one-half. The

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Chairman's address. Read before the Section on Urology at the seventieth annual session of the California Medical Association, Del Monte, May 5–8, 1941.

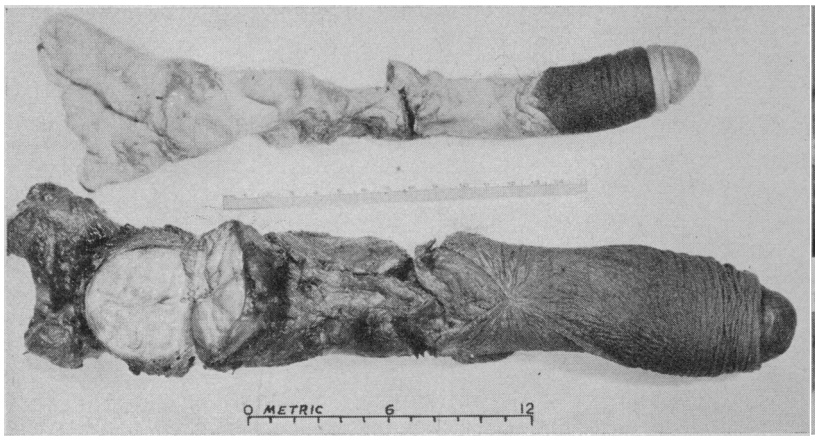

Fig. 1.—Autopsy specimen of the penis invaded with carcinoma, with normal control.

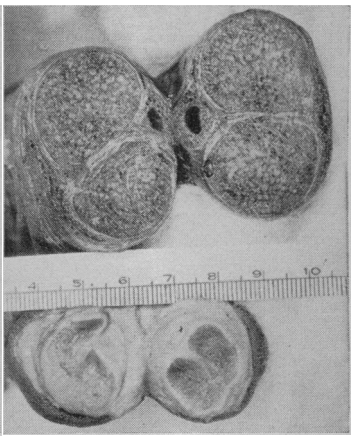

Fig. 2.—Cross section of the penis, showing invasion of the erectile tissue, with normal control.

urine obtained by ureteral catheter from the right side was normal, while that from the left was grossly bloody. B. coli were cultured from this kidney.

The pyelograms, made with Skiodan, showed the right kidney displaced downward, but the pelvic outline essentially normal. In the left kidney, the upper and middle calyces were distorted, and the diagnosis was made of a tumor of the upper pole of the left kidney. We felt that possibly the displacement of the right kidney might be due to external inflammatory disease, possibly an old perirenal infection, which would also account for his prolonged severe illness during the summer. The bleeding left kidney was lavaged with silver nitrate several times in an attempt to stop the bleeding.

A transfusion was given the next day, but the bleeding continued, so a left nephrectomy was done four days later. A large solid tumor was found, involving the upper pole, extending into the perirenal tissue and the pedicle, so that the nephrectomy was very difficult, and we did not feel that the tumor was completely removed. There was invasion of the renal vein and we encountered a good deal of bleeding. He was given one transfusion on the table and another later in the day.

The postoperative course was rather stormy, but there were no serious complications and he was able to leave the hospital on the nineteenth day. However, about ten days after the operation, approximately two weeks after the cystoscopy, while still in the hospital, he began to complain of tenderness and soreness in the penis. Examination revealed a diffuse swelling of the distal half of both cavernous bodies, without apparent involvement of the glans nor of the corpus spongiosum. From that time until his death, two months later, the penis gradually became larger and he continued to complain of tenderness and soreness, but never of any urinary difficulty. There were no nodules at any time, but the diffuse swelling increased and gradually included more of the organ. There was never an erect position, but the penis became about four times the normal size. The clinical diagnosis was made of thrombosis of the cavernous bodies, probably due to metastatic cancer.

Death occurred on November 28, 1938, from malignant cachexia and termnial bronchopneumonia. The autopsy was done by Dr. A. G. Foord, pathologist of the Huntington Hospital, Pasadena, who had also examined the operative specimen.

I have summarized the important findings of Doctor Foord's rather extensive descriptions.

The primary tumor involved nearly the entire upper pole of the left kidney, had invaded the renal vein, and had broken through into the perirenal tissue. It was only partially encapsulated, and had the typical gross yellowish appearance of a clear-cell adenocarcinoma, or so-called hypernephroma. The histology was typical of this tumor. There was a fairly well defined capsule, which was, however, extensively invaded. In the stroma of the capsule, and particularly growing in the blood vessels, including the renal vein, were solid nonvacuolated tumor cells, showing no evidence of foamy cytoplasm. Numerous veins were distended by nests of these cells with abundant solid staining cytoplasm and large hyperchromatic nuclei. There were a few nests of these cells throughout the tumor, but they were most abundant in the blood vessels and lymphatics of the tumor wall. It will be seen, therefore, that there were two distinct types of cells in the tumor, though Doctor Foord called it an adenocarcinoma, or hypernephroma.

The important autopsy findings were:

1. Local recurrence in the form of a mass involving the left adrenal and the pedicle of the kidney.
2. Microscopic metastases in the liver and lungs, without gross evidence of tumor in these organs.
3. Solitary metastases in the tail of the pancreas and in the substance of the right kidney.
4. A solid metastatic globular mass, about five inches in diameter, above the right kidney, attached retroperitoneally to the under surface of the liver. It completely surrounded and compressed the right adrenal, without invading it. This mass obviously was responsible for the low position of the kidney shown in the pyelogram.
5. A small encapsulated abscess lateral to the cecum in the lumbar gutter. This apparently was the cause of his prolonged illness and fever before we saw him, but no connection with the bowel or the appendix could be demonstrated.
6. The penis was profoundly enlarged, measuring in the embalmed state 23 centimeters from the apex of the prostate to the meatus, with an average diameter of 5 centimeters. The glans was not involved, but the distal 7 centimeters of the shaft was particularly large and firm, and the whole organ was in a state of erection. Multiple sections show marked enlargement of both corpora cavernosa, with distension of the cavernous spaces, some being 2 to 3 millimeters in diameter, with the lumen filled with grayish tan material, apparently tumor growth. At the base of the penis the corpus spongiosum was slightly invaded, but the urethral channel was fully 5 millimeters in diameter and not obstructed. The deep dorsal vein and several smaller tributaries were plugged with a gray-red thrombus, which extended upward into the periprostatic plexus. The left pudendal vein and the internal iliac vein were also thrombosed to the level of the junction with the external iliac vein.

The histology of the metastases showed two distinct features. The first was that, whereas the primary tumor was composed largely of the typical, large foam-cells characteristic of hypernephroma, with only groups of polyhedral solid staining cells especially about the periphery and in the lumen of the vessels, it was these latter cells which made up the greater part of the metastases and even the local recurrence. The large mass surrounding the right adrenal was made up of the typical foam-cells, but in the other metastases practically no cells of that type were found. The second feature was that everywhere the metastatic tumor cells seemed to select the lumen of the vessels, with relatively slight invasion of the stroma of the organ

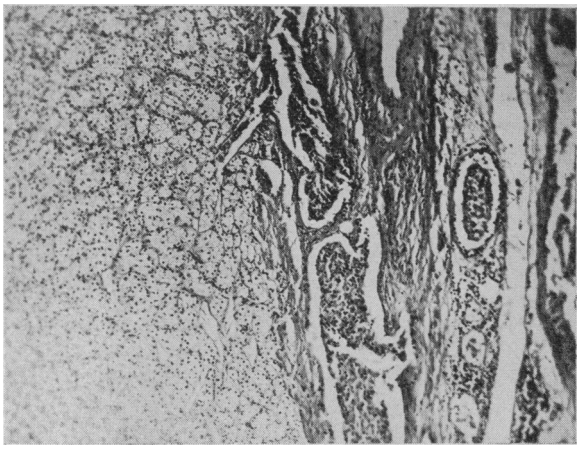

Fig. 3.—Section of the primary tumor, showing invasion of the venous spaces of the capsule with a different type of tumor cell from the typical hypernephroma.

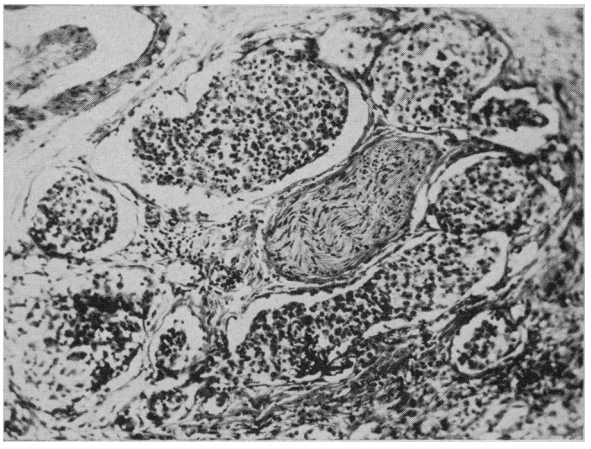

Fig. 4.—Microphotograph of the metastatic tumor of the cavernous bodies, showing tumor cells filling the venous spaces.

involved. This was especially noticeable in sections through the cavernous bodies, which showed marked distension of the venous channels, completely filled by tumor cells. Most of the growth was necrotic, but in the smaller channels viable tumor was seen, composed entirely of the solid type of cell, and without invading the stroma.

DISCUSSION

The rarity of this condition is shown by the fact that Begg,[12] in 1928, reported what is apparently the only similar case in the literature, a patient with metastatic hypernephroma in the cavernous bodies. His patient was a man of 62 who complained of a persistent priapism for five weeks. He had a greatly enlarged prostate by rectal examination. Cystoscopy being impossible, a suprapubic enucleation of the prostate was done, hoping to reduce the priapism. The patient died, and the autopsy revealed a large hypernephroma of the left kidney, entirely unsuspected during life. The corpora cavernosa were found densely infiltrated with typical masses of tumor, but the capsule was not involved. In this case, strangely enough, no other metastases were found in the body.

While preparing this paper, Dr. G. S. Sharp, consultant to the neoplastic clinic at the Veterans' Hospital at West Los Angeles, gave me the following incomplete account of a patient seen there last summer. This man, 45 years of age, had a right nephrectomy for tumor some five years ago. He did well for five years, then began to develop recently a constant priapism. General examination was negative, and x-rays of the chest showed no metastases. The penis was enlarged and erect, measuring 16 centimeters long, and 5 centimeters in diameter. There was one ulcerated nodule on the glans, about 1 centimeter in diameter. Of eight other definite nodules along the shaft, two were ulcerated. No biopsy was done, but a clinical diagnosis of metastatic carcinoma was made and x-ray therapy advised. The patient left the hospital and returned to his home in Salt Lake City, where I understand he is still alive.

The most common site of a primary malignancy from which metastases to the cavernous bodies occur is the prostate. I have succeeded in finding nine such cases. However, the prostate is an adjacent organ, and it must be realized that in any or all of these cases the involvement of the cavernous bodies may be by direct extension. Young says that carcinoma of the prostate often invades the walls of the membranous urethra, presumably through the lymphatics, and less often pushes on into the bulb. When this occurs, the erectile tissue is invaded, the sinuses being filled with tumor cells. At the base of the penis the process may break into the corpora cavernosa and eventually produce a carcinomatous priapism. The great numerical preponderance of these cases supports such a view.

OTHER REPORTS

Two of the primary tumors of the prostate were sarcomata. One, reported by Cowie,[13] was a boy of nine who had a priapism for several months. The autopsy by Warthin revealed an "oat-cell" myxosarcoma, primary in the region of the prostate, with extensive metastases in the cavernous bodies and elsewhere. The other sarcoma was found by Chauvin and Emperaire [14] in a boy of 20. He had an incomplete priapism from several metastatic nodules in the erectile tissue. X-ray therapy was used as palliative treatment. I have excluded the sarcoma described by Frantz and Alyea, though it has been referred to as of prostatic origin.

The seven examples of carcinoma of the prostate were reported by Toffier,[15] Nogues,[16] Guibal and Pavie,[17] Paglieri and Schiappapietra,[18] Young,[6] and Cecil.[19] Cecil has seen two cases, one of which he reported to me by personal communication. Young felt that his case can be explained as a direct extension, and this is possibly true of the others. Paglieri and Schiappapietra made a positive diagnosis by aspiration of the prostate through the perineum for biopsy, and also of a nodule in one of the cavernous bodies. Guibal and Pavie treated their patient by radium and x-ray, followed by complete emasculation, but do not record the outcome. All the others were terminal cases.

There were two primary carcinomas of the bladder. One was the patient of Neumann, mentioned by both Scheuer and Hinman. This patient, a man of 50, had a cancer of the base of the bladder which

penetrated through the wall. Autopsy revealed cancer invasion of all the veins of the penis and of the cavernous spaces. Priapism was due to an associated cavernitis. Kessell,[20] in 1934, described a malignant papilloma in a man of 39, with extensive metastases, including nodules in the cavernous bodies, with histology identical with the primary tumor. Kessell was able to observe the development of the priapism during the course of the disease.*

Three examples of metastases from primary malignant tumors of the testicle have been published by Froin and Pignot,[21] Bergeret,[22] and Garofalo.[23] In all of these the tumor seems to have been of the seminoma type, and there were extensive generalized metastases. The patients were respectively 52, 31, and 35 years of age.

Gadrat [24] has described the only patient in whom the primary tumor was in the liver. The man was 52 years old and complained of an enlargement and thickening of the penis, with partial erection. Gadrat used the term "false priapism." He had a tremendous liver, filling most of the abdomen. Diagnosis was made by biopsy puncture of the penis, which revealed malignant cells histologically typical of primary cancer of the liver.

As a point of interest, Guibal and Pavie also describe a squamous cell cancer of the cervix, treated by radium, with later metastases in the cavernous bodies of the clitoris. These two were given radium therapy, and subsequently the clitoris was removed. At the time of the report the woman was apparently well. The same authors also mention a cancer of the ampulla of the rectum, with metastases to the bulb and perineal portion of the corpus spongiosum, without invasion of the penis.

SUMMARY

Excluding these last two cases, which do not belong in the series, we have, including our own, eighteen cases of metastatic tumor of the cavernous bodies, of which nine were primary in the prostate, and subject to the criticism already mentioned. Of the remaining nine, all but one were primary in the genito-urinary tract. From the bladder the involvement may have been direct or by either retrograde lymphatic or venous extension. In the case of the more distant tumors, it seems most logical to assume a hematogenous route, especially since, in nearly every case, there were extensive general metastases, including the lungs. The three writers who described the tumors of the testicle all seemed to accept this explanation. An exception is the hypernephroma described by Begg, in which there were no other metastases. Why a tumor should select the cavernous bodies to the exclusion of all other sites is a mystery.

As already mentioned, the priapism was usually incomplete, and with the exception of the prostate cases, the patients were usually able to urinate with some difficulty. Usually the erection came on spontaneously before the patient was seen, but occasionally there had been an operation on the primary tumor, and in one case radium had been implanted into the prostate. In one patient the priapism followed immediately after a cystoscopic examination, and in several there had been such examinations. In my own patient the penis was apparently normal two weeks before the enlargement was noticed, when the man was cystoscoped. Usually the first invasion was in the form of discrete nodules which gradually coalesced. In my own, and in the other case of hypernephroma described by Begg, the involvement was diffuse from the onset. The relative rapidity of growth has been noticed by practically all who observed the condition in its early stages.

Accurate diagnosis was made in two instances by biopsy puncture which revealed malignant tissue in the cavernous body sufficient to make such a diagnosis. In both these cases it confirmed what was already a very strong clinical diagnosis of malignancy. However, since aspiration has been suggested by McKay and Colston [25] as a treatment for priapism, there would seem to be no objection to its use as a diagnostic procedure. Since nearly all the cases were terminal, the only treatment was palliative as a rule, usually deep x-ray to the penis, and in the prostate cases suprapubic cystotomy for the relief of obstruction. In the one case in which complete emasculation was done by Guibal and Pavie, the outcome was not stated.

IN CONCLUSION

The subject is presented on account of its unusual nature and pathological interest. In my own patient the sequence of events made a presumptive diagnosis easy without biopsy, but in doubtful cases the aspiration-trocar method would seem indicated. Despite its rarity, metastatic carcinoma must be considered as a cause of enlargement of the penis and priapism.

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* A third case of metastatic carcinoma in the erectile tissue, from a primary tumor of the bladder, was reported by Kreutzmann[26] in 1938.

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SICKNESS INSURANCE AND HEALTH SERVICE: A DIFFERENCE

WITH SPECIAL REFERENCE TO CALIFORNIA PHYSICIANS' SERVICE*

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THE job of making health service plans work has just begun. A great deal is known about hospitalization and it may be said that this phase is on a fairly accurate actuarial basis. This is probably responsible for the rapid growth of the Blue Cross hospital plan from 32,000 in 1932 to 6,300,000 in 1941. To show the speed with which the idea is taking hold, almost a third of the six million subscribers to hospital plans joined in 1940.

The medical phase of health service plans is a different matter. Until ten years ago the incidence of sickness was virtually unknown. Since then most of our information has come from surveys of one type or another. Most of these have been static in nature and have not shown sickness under conditions where it may be followed from day to day, week to week, month to month, and on. Furthermore, there are practically no usable statistics under health insurance systems adaptable to the American system and quality of medical care. Until such information is available, all designers of health service plans are following a path that offers few markers. California Physicians' Service is the only plan in the United States which is offering a "full medical coverage" on a state-wide basis and without any barriers between the physician and his patient.

CALIFORNIA PHYSICIANS' SERVICE

The weight of experience gained in California Physicians' Service may become all-important if and when the threat of State or Federal Government plans again become imminent. At present we may be having a breathing spell during which we may correct errors.

The 30,000 membership that California Physicians' Service now has is a small segment of the population and exposes each California Physicians' Service physician to an average of five potential patients. However, this membership is large enough to make the wheels of California Physicians' Service turn on a self-supporting basis and to provide experience in all the phases of a medical service plan from the problems revolving around the providing of medical service to individuals to the administrative problems related to care of large groups. There are many things that can and should be tried now with time to do sound planning and not under pressure of political encirclement.

* For news items concerning California Physicians' Service, see page 157.

APRIL TABULATIONS

An analysis of figures for the month of April, 1941, has been made showing the percentage of beneficiary members in each of the twenty-one California Physicians' Service districts, together with the percentage of units of professional service rendered in each district. Interestingly enough, the comparison discloses no wide discrepancies in extent and type of professional service. For example, in Los Angeles and Alameda counties (communities which have almost the same number of members) the cost of medical service differs only one per cent. A comparison on an over-all basis shows that the northern part of the state, with 63.6 per cent of the membership, has used 60.8 per cent of the units of service, while the south with 36.4 per cent of the membership has used 39.2 per cent.

ANALYSIS

According to April figures, the physician-patient load is rather evenly distributed throughout the state, with the single exception of Alameda County, where 6,498 members are served by 547 physicians. The state-wide average is less than four patients per year per physician.

The most important, and perhaps the most alarming, fact that is showing up in studies being conducted in the California Physicians' Service office, is the high incidence of illness. This has been consistently over 17 per cent in the past seven months. A high incidence was expected in the winter months, but it should begin to taper off in February. Instead the high figure for December, an epidemic month, has maintained itself into April. The epidemic is certainly long over, so it cannot be blamed. It is hardly conceivable that one out of every five of the beneficiary members required medical care consistently over a period of seven months. Yet these are the facts as recorded on the California Physicians' Service statistical machines. This extraordinary use of service is naturally burdensome to the whole plan. If the incidence of use of service could remain consistently around 10 or 12 per cent rather than at the present 17 to 19 per cent, California Physicians' Service would very quickly show a very different picture.

The record shows also that a major proportion of California Physicians' Service patients are being treated for minor chronic diseases. That 17 cents of each California Physicians' Service dollar is spent for x-ray and laboratory indicates that physicians are doing very thorough diagnostic work, which is essential to the practice of good medicine. However, this type of diagnostic work is apparently revealing pathology that necessitates continued treatment in a large number of cases. This seems to be borne out by figures showing that the same patients are receiving care month after month.

There are an unhealthy number of cases (from the standpoint of both California Physicians' Service and the health of the community) that continue on indefinitely. It may be that patients are demanding too much service for too many minor things, or it may be that too many doctors are finding too many things wrong with too many people. These